CASE REPORTS

Prolonged Survival of Schistosoma Japonicum

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The life-span of schistosomes in the human host is a poorly understood aspect of bilharziasis. Because of the possibility of reinfection, the longevity of the parasites cannot be reliably determined in untreated patients living in endemic areas. A few reports in the literature describe patients who were excreting living schistosome ova 20 years or more after leaving an endemic region. In most cases the ova were Schistosoma haematobium and Schistosoma mansoni. A patient with living Schistosoma japonicum seen in rectal biopsy 47 years after emigrating to the United States is reported herein.

Severe and often fatal liver damage is not uncommon in endemic areas where repeated and massive infections with S. japonicum occur. However, to have acquired the infection in youth, moved to a nonendemic area where repeated infection is impossible, and then to have suffered severe and eventually fatal liver damage from deposition of ova by adult flukes over a 47-year period is definitely unusual.

Report of a Case

A 65-year-old retired cook was admitted to the San Francisco General Hospital on September 30, 1965, because of abdominal swelling for two months. He also complained of ankle edema, a dull abdominal pain, and generalized loss of strength associated with a 15-pound loss in weight. He denied excessive intake of alcohol, previous jaundice, exposure to hepatotoxins, or exposure to patients with hepatitis. The patient emigrated to California from the Philippines in 1918 and has not been out of the United States since.

On physical examination the patient was thin and emaciated, and the abdomen was greatly distended. The blood pressure was 150/70 mm of mercury, the pulse rate 88 and respirations 18 per minute and the temperature normal. The skin was not icteric and no spider angiomas were seen. The abdomen was tense and there was an obvious fluid wave. The liver and spleen were not palpable. There was slight pretibial edema, but no clubbing or asterixis.

Laboratory data included hemoglobin of 13.1 grams per 100 ml and a white blood cell count of 5,500 per cu mm with a normal differential. Serum electrolytes, creatinine and blood glucose levels were within normal limits. Total bilirubin was 3.8 mg per 100 ml (normal less than 1.4), serum alkaline phosphatase of 3.4 Bessey-Lowry units (normal less than 3.0 units), serum glutamic oxaloacetic transaminase 50 Frankel units (normal less than 40 units), and total serum protein 7.2 grams per 100 ml with 2.4 grams albumin. Prothrombin time was 44 percent, and serologic tests for syphilis were nonreactive. Sulfobromophthalein retention was 24 percent. Stool specimens were negative for ova and parasites.

A liver scan showed the liver was small and the areas of decreased uptake widespread. Abdominal paracentesis yielded cloudy yellow fluid with a specific gravity of 1.015 and protein concentration of 1 gram per 100 ml. The cell block of the ascitic fluid was Class I. On November 19, peritoneoscopy was performed. The liver was small and diffusely and coarsely nodular and the spleen was moderately enlarged, but there was little venous enlargement of the other viscera. Biopsy of a specimen of liver obtained under di-

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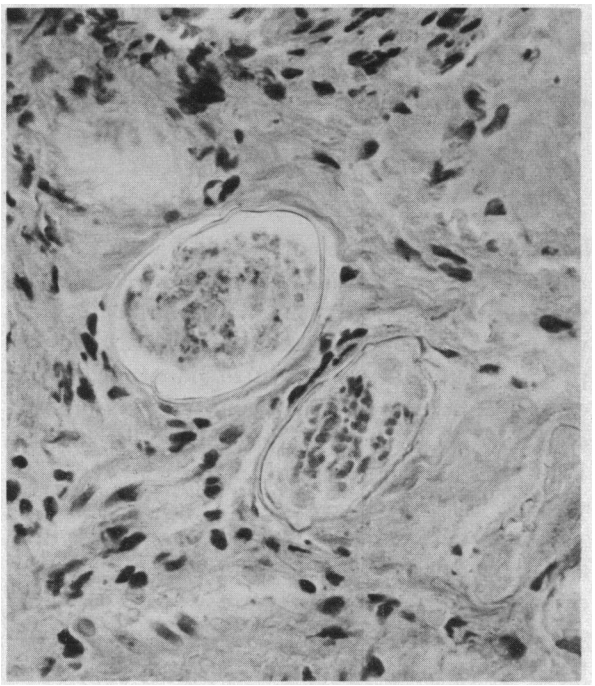

Figure 1.—Liver biopsy specimen showing schistosome ova within a decidedly distorted portal area. (Hematoxylin-eosin stain, X 400.)

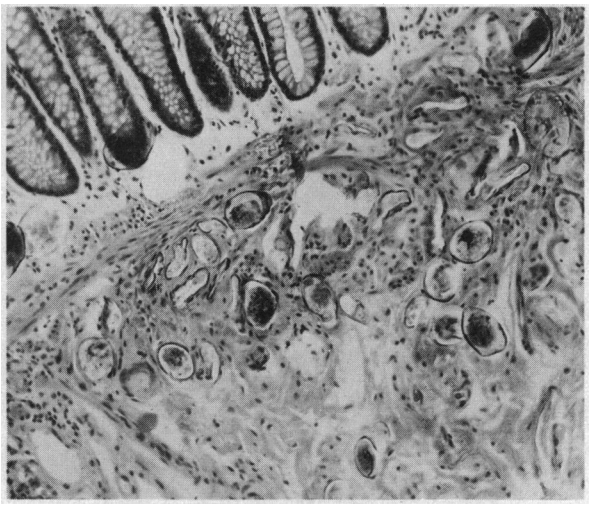

Figure 2.—Rectal biopsy specimen with multiple ova visible in the mucosa and submucosa. (Hematoxylin-eosin stain, X 100.)

rect visualization revealed pronounced portal fibrosis with focal bile duct proliferation and a mononuclear inflammatory infiltrate. Parasitic ova characteristic of S. japonicum were present within the areas of portal scarring (Figure 1). Subsequently, rectal biopsy showed numerous ova in the mucosa and submucosa (Figure 2). Examination of a fresh specimen revealed the oval-shaped living ova with tiny lateral spines. Flame cell movement was seen in some of the ova.

On December 21, 1965, catheterization of the right side of the heart revealed normal pulmonary artery pressure and pulmonary vascular resistance, but the hepatic vein wedge pressure was 17 mm of mercury (normal is 5 to 6 mm). During the three-month stay in hospital diuretic therapy was carried out with mercurial and thiazide preparations and spironolactone, and restriction of sodium and water. Intravenous tartar emetic therapy was begun January 1, 1966, with frequent electrocardiographic monitoring. Therapy was discontinued prematurely because of the development of early signs of nephrotoxicity (red cell casts in the urine and mild elevation of the serum creatinine). The patient had lost approximately 40 pounds in body weight by February 2, 1966, and although liver function tests were still mildly abnormal and the spleen decidedly enlarged, he was discharged.

When the patient was last seen in June 1967, ascites had reaccumulated despite maintenance diuretic therapy. He died several months later.

Comment

This case is unusual in that the clinical signs of S. japonicum infection became manifest 47 years after the patient left an endemic area. Since the parasite is not endemic in the United States and the patient had not traveled outside the country, it appears certain that he became infected during childhood in the Philippines where schistosomiasis due to S. japonicum is endemic. Although calcified and dead ova may persist in the liver for many years after the adult worms have died, the presence of viable ova in the rectum in this patient indicates the persistence of living worms. The presence of severe liver disease with elevated hepatic vein wedge pressure is in contrast to the classic "pipe stem" fibrosis seen in hepatic schistosomiasis, but it represents a well documented form of the late stages of liver involvement.[1]

In brief, the life-span of the parasite is as follows: 1) the human is infected, 2) eggs are excreted and 3) deposited on the ground, 4) rain washes the eggs into fresh water streams, 5) in the water eggs hatch into miracidia, 6) miracidia

penetrate the body of susceptible snails, and 7) undergo several stages of development in the snail, 8) the emerging cercaria (with tail) penetrate skin or mucous membranes of man, 9) make their way to liver where they mature into adult worms, 10) leave the liver, 11) remain in small venules of bowel wall where the females lay multitudinous eggs, which 12) are carried to all parts of the body, particularly the liver, brain and lung, and 14) deposition of ova in the tissues gives rise to granulomatous lesions and fibrosis.

When confronted with liver disease in persons from the Orient, one should consider the possibility of infection by S. japonicum. The mean lifespan of S. japonicum has been estimated at four and a half years by Hairston,[2] while Belding[3] reports a longevity of 20 to 25 years. A few case reports document prolonged survival of the schistosomes in patients who have migrated to nonendemic areas. Berberian, Paquin, and Fantauzzi[4] reported a case of infection with both S. mansoni and S. haematobium by a Yemenite man who had been living in New York State for 27 years. Wallerstein[5] described viable ova of S. mansoni, found on biopsy of a rectal polyp in a Puerto Rican woman after she had lived in New York City for 26 years.

The case reported herein documents an unusually long survival of S. japonicum in a patient removed from an endemic area, and suggests that the lifespan of the worms in untreated human hosts may be very long.

Summary

A case of Schistosoma japonicum infection with severe hepatic involvement and living ova in the rectum in a patient who emigrated from the Philippines 47 years previously is presented. Since the parasite is not endemic in the United States, this represents another example of the previously reported long survival of schistosomes in the human host.

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Retroperitoneal Fibrosis in Two Patients Previously Exposed to LSD

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IN MOST CASES of retroperitoneal fibrosis the cause is obscure.[1] A minority of cases are due to methysergide maleate.[2] Other drugs which have come under suspicion include ergotamine, dihydroergotamine, hydralazine,[2] sulfonamide[3] and tetracycline and penicillin.[4] Most patients, however, have no history of drug exposure. Ormond[5] is amongst those who feel that allergens may eventually be found to be the cause, but there is as yet no proof that methysergide causes the disease through an immunologic mechanism rather than because of its pharmacologic effects. Graham and his colleagues[2] have suggested a number of ways in which methysergide might cause the disease.

Two patients seen at the Stanford University Medical Center because of retroperitoneal fibrosis gave a history of past exposure to lysergic acid diethylamide (LSD). Although there was no clear evidence that LSD contributed to the disease in these two patients, the occurrence is worthy of note because of the chemical and pharmacological similarity of LSD to methysergide (Figure 1).

Reports of Cases

Case 1. A 42-year-old man had labile hypertension, noted at an Armed Services physical examination at 18 years of age. In the ensuing years his diastolic blood pressure had been between 85 and 90 mm of mercury as determined

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